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Simultaneous presentation of three types of osteoid osteoma in the maxilla

KEYWORDS
Osteoid osteoma;
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Osteoid osteoma (OO) is a benign osteoblastic bone tumor that predominantly affects young adults and has a higher prevalence in men.[1] OO accounts for approximately 10 %–14 % of all benign bone tumors and 2 %–3 % of all bone tumors overall.[2] OO typically presents as a solitary lesion in the appendicular skeleton, but when it occurs in the jaws, it is most commonly found in the mandible.[3] A hallmark symptom is nocturnal pain that responds well to nonsteroidal anti-inflammatory drugs. Radiographically, OO is characterized by a radiolucent nidus, usually less than 1 cm in diameter, with central calcification, surrounded by a zone of sclerotic bone or cortical thickening.[4] The tumor is classified into four subtypes based on location, namely subperiosteal, cortical, endosteal, and medullary, with most lesions situated in or near the cortex.[4] In the jaws, the medullary subtype may be misdiagnosed as an odontoma due to overlapping features. Herein, we reported a rare case of multifocal OO of the maxilla in an older female patient.

A 61-year-old female patient was referred to our oral and maxillofacial surgery department for evaluation of multiple painless mixed radiopaque and radiolucent lesions in the left maxilla. Cone-beam computed tomography (CBCT) revealed three primary lesions (Fig. 1A and B): the most superior lesion was located at the level of the left nasopyriform region (Fig. 1C), the second lesion was situated a few millimeters below the first (Fig. 1D), and several additional lesions were observed between the roots of the left maxillary canine and first premolar (Fig. 1E). Base on the lesions' radiographic appearance and clinical presentation, the OO, with subtypes corresponding to cortical, endosteal, and medullary forms were suspected. The differential diagnosis for the two superior lesions included osteoblastoma, whereas the medullary lesion raised the possibility of an odontoma with cystic change (Fig. 1F).

Because of the patient's worry of malignancy, she requested surgical excision of the lesions. Under general anesthesia, the two superior lesions were readily excised (Fig. 1G). The lesions located between the roots were more challenging to excise because their margins were ill-defined and their characteristics differed from the odontoma with cystic change. To prevent damage to the adjacent tooth roots, a portion of the lesion was intentionally left in place. Histopathological examination confirmed the diagnosis of OO, consistent with the initial clinical impression (Fig. 1H–K). At the 1.5-year follow-up, the residual lesions remained stable, and the involved teeth retained normal vitality (Fig. 1L and M).

In this patient, although the two superior lesions could be readily diagnosed as OO on the basis of CBCT findings, the multifocal presentation is atypical for OO. Moreover, the absence of pain, a hallmark feature of OO, limited the clinical utility of symptomatology in differentiating OO from osteoblastoma. In addition, the curved morphology of the lesions between the roots closely resembled that of an odontoma with cystic change, which could not be definitively ruled out preoperatively. Although surgery is invasive, obtaining tissue for pathological examination remains the only method to diagnose this atypical case.

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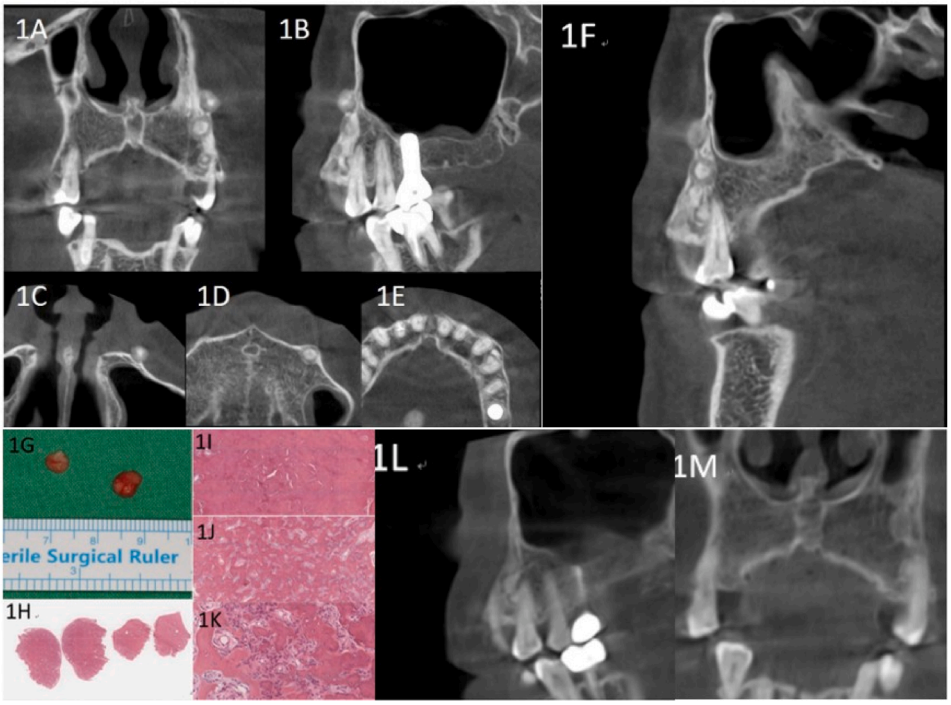

Figure 1 Clinical photographs, histological photomicrographs, and radiographs of the case. Preoperative cone-beam computed tomography (CBCT) of the patient: (A) Coronal view, (B) sagittal view, (C—E) axial views depicting three mixed radiopaque and radiolucent lesions in the left maxilla. (F) Sagittal view of preoperative CBCT illustrating an odontoma-like lesion near the root of the left premolar. (G) Gross image of the two excised specimens. (H) Gross histopathologic view of the lesion and (I—K) photomicrographs at 40 × , 100 × , and 400 × magnification, respectively. Microscopically, the lesion was well defined, featuring centrally located thick sclerotic bone and peripherally located lace-like, anastomosing trabeculae of woven bone. Prominent osteoblastic rimming was observed. Postoperative CBCT of the patient: (L) Coronal view, (M) sagittal view showing residual medullary-type osteoid osteoma between the left maxillary canine and first premolar.

Declaration of competing interest

The authors have no conflicts of interest relevant to this article.

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